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STOMATITIS NEUROTICA CHRONICA.

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STOMATITIS NEUROTICA CHRONICA.¹

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OF NEW YORK.

CASE I.—C., fruit business, forty-two years old, married; had one child which was stillborn. His father is alive, seventy-eight years old; his mother seventy-five. There are no other relatives except one sister, who is described as being perfectly normal, and one brother, whom I personally know. He appears fairly well developed physically, but has no whiskers, no hair on the pubes, none in the axilla, and the sexual organs are those of a child of ten years. Both of these persons have had pemphigus in the mouth all their lives.

Patient appears to be pretty well developed, his complexion, though, is sallow; all his internal organs are in fair condition. His urine has a specific gravity of from 1.018 to 1.025; is always negative. He has always complained of a sensation of fatigue. He tires very easily, drags his limbs; cohabitation, which occurs but once a month, exhausts him; his hands were always weak, and moderate exertion results in tremor. He has trouble in keeping warm, and complains a good deal of præcordial pain, both on pressure and before his meals. This pain, as he expresses himself, was never so bad as when, under the orders of a physician, his diet was restricted. He has a melancholy temperament, and expresses now and then a number of hypochondriacal notions.

Up to his thirteenth year he suffered severely from urticaria. Since his thirteenth year this urticaria has disappeared, and has been replaced by pemphigus in the mouth. A few blisters will form quite suddenly, sometimes on the tongue, mostly on the cheeks, frequently in their lower parts, near the alveolar processes. They will burst after a little while; whitish ulcers will remain quite a long time and give rise to a great deal of pain,

¹ While this paper was being put in print, Dr. O. Rosenbach published (*Deutsche med. Wochens.*, June 28, 1894) a paper containing a "Contribution to the Vesiculating Affections of the Oral Mucous Membrane." Its conclusions are as follows: "The local pemphigus of the mucous membrane of the mouth is a variety of erythema exudativum multiforme, and ought to be called erythema bullosum. Whatever has been described as urticaria, herpes, etc., of the oral mucous membrane, is mostly this erythema bullosum. It is often complicated with the same eruption on the genital organs. It has a tendency to relapse."

The very variety of the symptoms, and the etiological identity evidenced in the histories of the cases, makes me believe that the name selected by me is more significant and appropriate.

sometimes resulting in sleeplessness. Half of his lifetime, since his thirteenth year, he has been troubled with this eruption, which would come on without any apparent cause. He states that half of this period he has been free, not being able to explain why he should have the eruption nor why he should have been free.

About ten years ago, after having been under treatment three years, he had no eruption during the course of three or four years. At that time, and in some later periods, the medicinal treatment consisted of the internal administration of atropia, arsenic, and ergot. He claims that no local treatment ever benefited him except the application of concentrated sulphuric acid. He says it was a notion of his own, and asserts that it heals the sores in a few days.

Once, when the patient was as willing as I had been long ago, I sent him to a neurologist of well-merited fame and sound judgment. What he replied was as follows: "I could not make out the cause of the ulcers in C.'s mouth. If it is not a state due to chronic indigestion, and yielding to it, there are two possibilities to consider—one, that he is an epileptic; that the sores are due to attacks in which he bites his cheeks and lips, and of which he is wholly unconscious; secondly, that a morbid tendency leads him to use various acids, etc., in his mouth (and this he admitted he did), which keep up the ulceration or reproduce it. He seemed to me a queer hypochondriacal person, and gave me such an indefinite history that I suspected some mental taint."

CASE II.—Mrs. W., thirty-two years old, the oldest of five children in the family. Her father has a very irritable, choleric temperament; her mother is placid, a woman of character and intellect. Her oldest brother has always had a systolic heart murmur, without any hypertrophy, the cause of which can be referred only to an insufficient contraction of the heart muscle. He is now a man of twenty-eight. He has for the last six years had peculiar attacks (epileptic?), the principal symptoms of which are slight contraction of the sterno-cleido-mastoid, which turns his head to the right side; scintillation and semi-unconsciousness, which is sometimes complicated with slight delirium, temporary forgetfulness, and limitation of his field of vision on both sides. These attacks will sometimes come on every month, sometimes not in six months. They are frequently the result of sudden turning of his head in one direction, and upward.

Another brother had meningitis when three years old, with all the symptoms of a tubercular meningitis. The disease did not prove fatal, but from that time on the boy had a feeble intellect, amounting to stupidity; was a bad scholar at school; had an irritable, violent temper, amounting to moral insanity when quite young; became more violent when he grew up, and had finally to be confined in an asylum for the insane, where he has been these many years.

A sister of the patient has suffered from petit mal since childhood, and has feeble intellect, still she is married. A younger sister died when five years old, of convulsions during pneumonia.

Patient was always neurotic ; dysmenorrhœa has accompanied her through life ; she has been married six years, and has been pregnant four times. The first pregnancy resulted in abortion, the second in the birth of a boy who is four years old and suffers from dyspepsia and constipation. He is a very neurotic boy, cowardly, melancholic, and talks much of his health. The third and fourth pregnancies terminated in abortion.

The patient wore convex glasses, No. 12, twenty years ago ; wears No. 6 at the present time. She suffers much from headaches, which do not appear to depend on the condition of her sight. Cephalalgia will come on mostly about the time of menstruation, which is irregular, sometimes scanty, sometimes copious, mostly behind time. Headache will also be produced by reading, thinking, and when the stomach is empty, also under the influence of anxiety or emotions in general. During her headaches she is pale ; now and then, however, her forehead will be flushed. Her tendency to constipation will sometimes be interrupted by diarrhœa. Nausea is a frequent symptom ; it is often complicated with vertigo. Her kidneys are normal, the urine now and then quite pale and of very low specific gravity.

Other neurotic symptoms are, frequent thirst, also nausea when she stoops and when at work over drawers. In such cases vomiting has been observed which lasted a whole day. Music will confuse her. In the stores she will sometimes forget what she wished to purchase. Occasionally she complains of neuralgic sensations all over the face in every direction. She sees sparks. Whatever she does she complains she does mechanically. She has "to think terribly much."

Whenever she has such attacks of nausea, of vertigo, and during dysmenorrhœa she has a sudden eruption of vesicles in her mouth of a size from a lentil to a pea, preceded by local redness. They burst very soon and leave behind them a sore surface of grayish-white color, which feels a little harder than the normal neighborhood, the hardness evidently depending on a fibrinous exudation into the surface. It takes many weeks before these sores heal. Fortunately they are never numerous. In a number of instances only one would appear. She has had these attacks very frequently since her thirteenth year. Menstruation did not show itself before the fourteenth. The only times when she has been permanently free were during pregnancy and during a trans-Atlantic voyage some four years ago and an absence of about six months.

CASE III.—W., stationer, thirty-one years old ; never had eczema or other cutaneous diseases ; never a chill ; has regular bowels ; boasts of general good health, and has a great many local and general complaints. He is a good sleeper, his organs appear to be healthy, his urine has a specific gravity of 1.023 and is negative. He smokes but little, but both cigarettes and pipes. Boasts of not being a nervous man. His mother, however, was "quite nervous," and an only sister died of œdema glottidis at the age of thirty-one years. He has been addicted to masturbation from early life until a few years ago. For two years has had attacks of palpitation which last three

or four days, during which breathing is more or less difficult. His heart is of normal size, the sounds feeble; there is no murmur, no hypertrophy. He complains of his stomach, which has been bad three or four years; of slow digestion, and of attacks of vomiting some years ago. He eats rather fast, and takes alcoholic beverages from time to time. The gastric pain he complains of is in the median line. He has it mostly when the stomach is empty. The pain leaves him after eating and returns after an hour or two. His diet does not appear to influence this pain; as he expresses himself, he has pain after everything and is painless after everything.

When he presented himself in March, 1894, he gave the following history: During the last days of 1890 he was sleepless, from no apparent cause. He committed no indiscretion in his diet, but on the 1st of January, 1891, quite suddenly he had what he calls canker sores on his left cheek; blisters sprang up which opened soon and left a white surface which lasted weeks and gave him much pain. He says they travelled around the mouth on both sides, jumping over distances, and have not left him since with the exception of a few days about Thanksgiving Day in 1893. Until that time he had a large number of such blisters and ulcerations of the size of peas and less, rising from what appeared to be a normal surface, on lips and tongue, besides the cheeks. All at once, about Thanksgiving, 1893, he could again whistle and remained well a whole week. Then blisters would spring up again on the outside of the lips; the angles of the mouth cracked, and blisters appeared in the oral cavity, terminating in superficial excoriations. Since the 1st of December, 1893, when the new eruption sprang up, its character has somewhat changed. Besides the large pemphigus vesicles in his mouth, the outside of the lips has participated in the process. There the vesicles are smaller, and rise from a hyperæmic surface. The cheek will swell up now and then, and headaches are not of uncommon occurrence. On April 2d, he complained of a severe headache which had continued a week, and necessitated, and was eased by, prolonged sleep. This headache improved greatly since a few days previous to his call a frontal herpes showed itself. During that time his bowels and appetite had improved, but a symptom which had not been present before and staggered him much, was a very copious perspiration around his anus. On the 17th of May he reported this had disappeared; there was some pain on the right forehead; there was no pemphigus in his mouth, but a few pemphigus eruptions had appeared on the outside of his lips, which had lasted three weeks. He again makes the statement that he has not been free for a year, with the exception of that one intermission in November, 1893.

Pemphigus has amongst its causes heredity and certain affections of the nervous system. Kaposi¹ reports the case of a man of twenty-two years who suffered from pemphigus since his childhood.

¹ A. Eulenburg: Real. Encycl., 2d ed., vol. xx, p. 291.

His sister, his mother, her brother, and half of the children of the latter were similarly affected.

In anomalies of the sexual organs, in pregnancy (Hebra, Bulkley), after confinement (Köbner), it has been observed; in lepra (Boeck, Nielen) it appears either as prodromus and may last for years, or it originates—both traumatically and spontaneously—on anæsthetic parts of the surface. The spinal cord has been found to be altered in chronic pemphigus; it is met with in chronic myelitis and in neuritis. But in all of these cases pemphigus was not found on the mucous membrane, as in those related by me, and the following:

Kopp¹ describes a case of what he calls tropho-neurosis hysterica. It was that of a woman with a neurotic history and hysterical attacks. Amongst other eruptions she had herpes on the lower lip over the right mental nerve, and after that a small vesicle over the middle of the upper lip. This was followed by a large vesicle on the left side of the lower lip and a stomatitis with frequent hysterical attacks. Both vesicles and stomatitis disappeared in five days. Later on there was marked pain in the gums and on the mucous membrane of the mouth, followed by herpetic vesicles on the lower lip and a marked stomatitis, with fetid breath and frequent hysterical attacks. A week later there came another crop of herpetic vesicles on the lower lip, and the same condition would repeat itself at longer or shorter intervals. During all these attacks the temperature was normal. Alongside the vesicles there went a desquamating catarrh of the mucous membrane of the mouth, which recurred frequently and suddenly. Some months afterward, with a recurrence of the hysterical attacks and other skin affections, there was again herpes on the lips as before, and a marked stomatitis.

T. S. Flatau publishes,² under the head of chronic recurrent herpes of the oral cavity, the case of a merchant, thirty-eight years old, who was well until his eighteenth year. Then he had typhoid fever and became addicted to alcohol; it is thus that the neurotic element may be surmised to have come into the case. Some months after his convalescence from typhoid fever he had an herpetic eruption on the lower lip, which soon healed. Since that time he frequently had similar eruptions on the mucous membrane of the

¹ Trophoneurosen der Haut. Inaug. Diss., Würzburg, 1891.

² Deutsche med. Wochenschr., 1891.

lips, on the gums, near the floor and roof of the mouth, occasionally also on the tongue, mostly along the free margin posteriorly. These crops would last from eight to twenty-eight days. The longest time that he was ever free was one year, being then also relieved of the general irritability of his nervous system. Then the eruptions reappeared, and since that time the intervals between them have continually grown shorter. It was also observed that when his nose was clear there was no eruption, and when the latter appeared the nose was obstructed. He never had neuralgic pains, but salivation frequently. Patient shows marked mental depression, which he explains by the fact of his suffering from his trouble through so long a period and his impossibility to master it by treatment. There was no syphilis, no tuberculosis, no malaria. The simultaneous appearance of nasal symptoms might suggest a causal influence of the latter, but it is quite as probable that the mucous membrane of the mouth and of the nose was affected at the same time. But there is no means of arriving at a correct diagnosis of the original condition. The assumption of a peripheral or central affection of the trigeminus is hypothetical.

B. Mandelstamm¹ observed four cases of pemphigus of the mucous membrane of the mouth within ten years. In one of them the eruption was also seen part of the time on the skin. In all of them the treatment was unavailing. There was no systemic disturbance, but there was fetid breath and painful deglutition. Eruptions were found on the mucous membrane of the tongue, cheek, lips, and throat. The sores were of a size from a lentil to a cent. They were grayish-white or white. The patches had sharply defined borders. Some were thick, like a membrane, and could not be removed without leaving a bleeding surface. The eruptions, when on the tongue, were usually found on the free border of the inferior surface. Now and then they would appear on the epiglottis. Around the excoriations or membranes there was usually slight œdema, and salivation was not uncommon. Microscopic and bacteriological examinations were all negative.

The author has scanned the literature quite carefully, and comes to the conclusion that such cases must be very rare. A little more

¹ Berl. klin. Wochenschr., 1891.

frequent are detachments of epithelium which remain membrane-like on the surface, and may resemble to a certain extent diphtheritic or croupous membranes.

With the exception of a few cases to be mentioned hereafter, the above cases of chronic pemphigus of the mouth appear to be all that have been observed. A few instances of pemphigus of the skin I now propose to mention briefly, and to accompany them with a very few remarks which are meant to elucidate the peculiar etiology of the affection.

Kirchner's case¹ was that of a man of fifty-four years, who was reported to have had a primary specific affection without, however, exhibiting secondary symptoms. For the past seven years he was frequently taken with pemphigus eruptions on arms, legs, face, and neck, accompanied with general systemic disturbance and sweating. Sudden exposures to cool air after perspiration were presumed to occasion the attacks. At the Ninth Congress² of Internal Medicine a few cases of pemphigus were the subjects of discussion. Mosler claimed it as a vasomotor, Schwimmer as a tropho-neurotic affection; as to its neurotic character there appeared to be no doubt. As the suspicion of its being of an infectious nature easily suggests itself, microbes were carefully looked for by Mosler, but not found. Nor was Landgraf more successful in this respect, in a case of chronic pemphigus of the conjunctivæ and the mucous membranes of the nose, fauces, and oral cavity.³ A similar case was reported by Boer⁴ and Chiari;⁵ it was observed in a coachman of sixty-three years, who developed pemphigus on the mucous membrane of the larynx. Tuberculosis was suspected, but the diagnosis was dropped when subsequently the same eruption showed itself on the throat and mouth. No attempt is made in the publication to diagnose the cause.

Mosler's case in a woman of thirty, of pemphigus of the skin of the trunk and extremities, was decidedly of a neurotic character, in a neurotic family. The patient was always of delicate health, and had repeated attacks of epistaxis, fainting, and epilepsy from her twelfth to her fifteenth year; afterward she suf-

¹ Archiv f. Dermatol. u. Syph., Wien, 1892.

² Verhandl., Wiesbaden, 1890, p. 252.

³ Berl. klin. Wochenschr., 1891.

⁴ Archiv f. Dermatol. u. Syph., 1890.

⁵ Wiener klin. Wochenschr., 1893.

ferred from chlorosis, dysmenorrhœa, arthritis, hæmoptysis, and hæmatemesis.

Benedict observed pemphigus both after peripheral nerve lesions and central disturbances. So have Chareot, Leyden, and Eulenburg. Hebra quotes Dickson and Gilibert, who claim emotional excitement as amongst the causes of pemphigus, and admits the existence of a "pemphigus hystericus" which appears and disappears in combination with a series of hysterical symptoms. A unilateral case of the kind is reported by Pick (*Wiener med. Presse*, 1880).

The majority of the neuroses of the integuments, which result either in a simple hyperæmia and œdematous tumefaction or in exudation reaching up to and into the epidermis, have been described as either urticaria or herpes. They are found on the skin almost exclusively. In severe generalized urticaria of the surface I have, however, repeatedly met with an œdematous swelling of the mucous membrane of the mouth; once I observed an acute laryngeal catarrh with croupous cough and dyspnœa, and once a very intense rectal tenesmus. Herpes febrilis I have seen quite often, when severe on lips and chin, to spread over tongue and cheeks, exhibiting the same character in all localities, and following the course of a peripheral branch of the trifacial nerve. As a rule, however, both urticaria and herpes are limited to the surface. Hemorrhages into the skin I have now and then found in exquisitely hysterical women. Pemphigus, as the literature of the subject proves, is but rare in that connection. Whenever found, it is on the skin almost exclusively, not on the mucous membrane. With the exception of the few cases given in detail, there are amongst the instances quoted above only those of Landgraf, Boer, and Chiari in which pemphigus was limited to the mucous membrane of the oral cavity.

Herpes is by no means rare; indeed, *acute herpes* ("zoster") is a frequent neurosis. Its form, however, is not always the same. In some eruptions of zoster the liquid exudation does not suffice for the formation of vesicles; in others the vesicles are very large, like those of pemphigus. These two varieties of dermatitis will sometimes resemble each other in many points. Mosler's case of cutaneous pemphigus began with small vesicles; within four or six hours they were of the size of pigeon's eggs. In one of my cases the pemphigus eruption was surrounded with a hyperæmic area, and that

of Kopp's is of a similar nature. These instances prove the at least occasional presence of equal etiological and pathological conditions. Still, the great searcity of pemphigus of the mucous membrane of the mouth appeared to me a sufficient reason both to describe my cases and to range them under a special heading. The neurotic character of my cases appeared to me self-evident. In two there is a positive family taint; in one the taint is but individual, but the neurosis finds its cause, amongst others, in masturbation, and exhibits a series of nerve anomalies. Even gastric symptoms, when they occurred in my cases, were easily recognized as neurotic. Other symptoms of the same nature were headache, constipation, diarrhoea, nausea, local perspiration, and general neurasthenia. The temperature of the body was always normal. Contrary to what we see in herpes of the surface, which heals readily unless maltreated, pemphigus of the mouth is of long standing, and heals but slowly, and gives rise to a great deal of protracted pain and discomfort.

The marked family disposition which is evident in the history of two of my cases, reminds me strongly of what Carl Blumer has described under the heading of "hereditary disposition to vesiculation" (Prague, 1892). Valentin employed for the same affection the name of hereditary dermatitis bullosa; Köbner called it epidermolysis bullosa hereditaria; and Klebs, dysplasia vasorum. He found the bloodvessels in a condition resembling that of hæmophilia; down to the capillaries they consisted pre-eminently of cells of embryonal structure, predisposing to hemorrhages and exudation. Some of the cases described by Blumer and others (Goldscheider, Valentin, Ferd. Hebra, and Köbner) were complicated with urticaria, swelling of the lymphatic vessels and lymph-bodies, tumefaction and redness of skin surrounding the vesicles, general malaise when eruptions were copious, hemorrhages into the vesicles, suppuration, and purpura.

